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[Case Report]

Generalized peritonitis secondary to spontaneously perforated pyometra in elderly women : two cases with different clinical courses and surgical approaches and review of the literature

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Abstract We describe two cases of spontaneously perforated pyometra (SPP) in elderly women treated with two different surgical approaches. An 88-year-old woman underwent emergency laparotomy for presumed diagnosis of gastrointestinal (GI) tract perforation. During surgery, SPP and a tumor of the sigmoid colon were identified. Total hysterectomy and sigmoid colon resection were performed. Despite exhaustive postoperative treatments, the patient died on postoperative day (POD) 189 due to peritonitis and pneumonia. A 93-year-old woman with acute abdomen was diagnosed with severe pyometra and primarily treated with transcervical drainage. Due to progression of generalized peritonitis, laparoscopic surgery was performed. Intraoperatively, scar from a uterine body perforation was identified, leading to the diagnosis of SPP. Only peritoneal irrigation and drainage were performed, in consideration of her advanced age. She improved and was discharged from the hospital on POD 35. The prognosis for SPP is sometimes poor, especially in older women. Minimally invasive surgical intervention might be considered for primary treatment in such cases.

Key words : spontaneously perforated pyometra, generalized peritonitis, diagnosis, surgical intervention, prognosis

Introduction

Pyometra is defined as an accumulation of purulent material within the uterine cavity. The incidence of pyometra is estimated to range from 0.1% to 0.5% in the general population, although it typically occurs in postmenopausal women[1,2)]. Spontaneously perforated pyometra (SPP) is very rare ; it is estimated to account for less than 5% of all cases of pyometra[1)]. Preoperative diagnosis of SPP is difficult. The prognosis is sometimes poor, especially in women of advanced age in poor general conditions. We present two cases of SPP and discussed preoperative diagnosis, surgical intervention, and prognosis with a review of the literature.

Case reports

Patient 1.

An 88-year-old woman with acute abdomen was emergently transferred to the department of surgery at our hospital. She complained of gradually worsening abdominal pain over 4 days. Of note, her general condition deteriorated so that she could not walk on the day of transfer. She had a history of hypertension and angina pectoris, both treated with medication. On examination, she had tachycardia (103 bpm) but normal blood pressure (105/57 mmHg) and no fever (35.1°C). Laboratory investigations revealed leukocytosis of 11,900 × 10^6/L and elevated level of C-reactive protein (CRP) at 16.29

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mg/dL. A preoperative gynecological examination was not performed due to her emergent presentation. She did not complain of any atypical genital bleeding or abnormal increase in vaginal discharge. She had not received any examinations for uterine cancer for over 20 years. Urgent computed tomography (CT) revealed a fluid collection and free air in the upper abdomen (Fig. 1). In the pelvic cavity, the uterine cavity was dilated with fluid and contained free air. Since generalized peritonitis due to gastrointestinal (GI) tract perforation was suspected, emergency laparotomy was performed. A midline incision in the lower abdomen, approximately 15 cm in length, was made during laparotomy ; purulent ascites was found in the entire peritoneal cavity. During peritoneal irrigation, surgeons found a perforation in the uterine fundus with purulent discharge leaking from the uterine cavity (Fig. 2A), which led to the diagnosis of SPP. Gynecologist was consulted ; the gynecology team performed a hysterectomy and bilateral salpingo-oophorectomy. Although there were no findings of GI tract perforation, surgeons found an abnormal mass that was strongly suspicious for malignancy in a segment of the sigmoid colon. Subsequently, surgeons resected the sigmoid colon. The operative time was 7 hours and 24 minutes ; intraoperative blood loss was 2,270 ml. Postoperative histopatho-

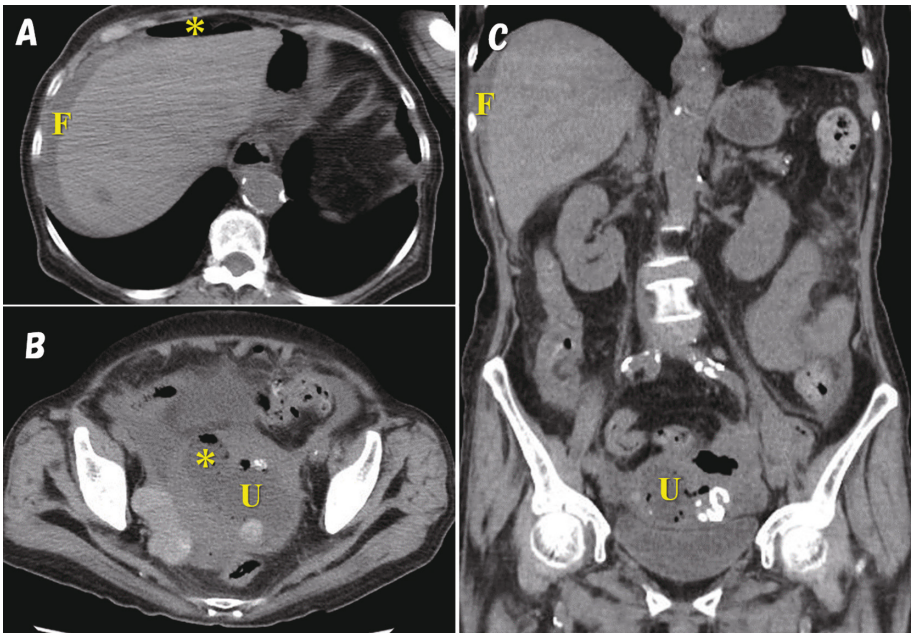

Fig. 1. Preoperative computed tomography (CT) findings in Patient 1.
Urgent preoperative CT revealed a fluid collection (F) and free air (*) in the upper abdomen (A). In the pelvic cavity, the uterine cavity was dilated with fluid and contained free air (A and B).

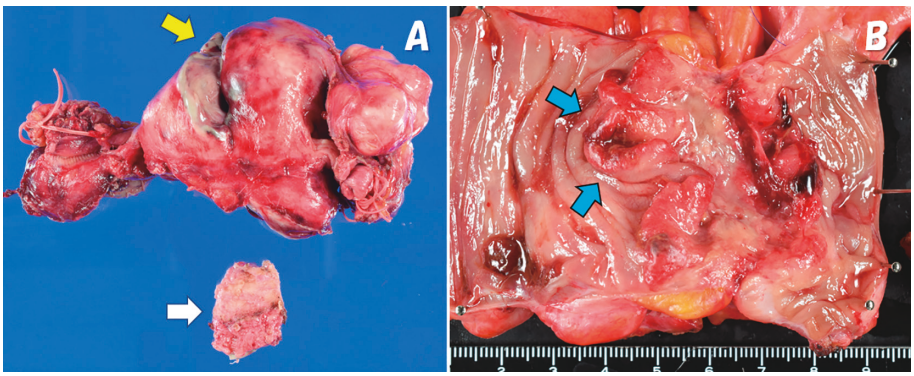

Fig. 2. Findings in the resected uterus and sigmoid colon in Patient 1.
The yellow arrow indicates the site of perforation in the uterine fundus (A).
The white arrow indicates the uterine cervix, which was friable during hysterectomy (A).
The bleu arrows indicate carcinoma of the sigmoid colon (B).

logical examination revealed leiomyoma and pyometra from the myometrium to the serosa, with no evidence of uterine malignancy but adenocarcinoma of the sigmoid colon (pT4a N0 M0 stage II) (Fig. 2B). Stenosis of the uterine cervix was noted. Culture of the peritoneal pus grew *E. coli*. Postoperatively, the patient received comprehensive treatment consisting of several antibiotics (CMZ, MEPM, IPM/CS, and LVFX were selected based on bacterial culture results from purulent material) in the intensive care unit. Diastasis of the sigmoid colon anastomosis occurred a month after surgery. Despite exhaustive treatments, the patient died on postoperative day (POD) 189 due to peritonitis and pneumonia that grew progressively worse.

Patient 2.

A 93-year-old woman was emergently transferred to the department of obstetrics and gynecology at our hospital with acute abdomen that began that morning. She had a history of hypertension and diabetes mellitus, both treated with medication. The patient needed assistance with activities of daily living because of left hemiplegia due to cerebral infarction 1 year earlier. On arrival at the hospital, her vital signs were stable. Laboratory investigations revealed leukocytosis of 11,900 × 10^6/L and elevated levels of CRP at 19.55 mg/dL. Urgent CT revealed a fluid collection and free air in an extremely dilated uterine cavity, although only a small amount of ascites was observed in the pelvic cavity (Fig. 3-A-C). The patient complained of abnormal purulent vaginal discharge, which lead to a diagnosis of severe pyometra. Transcervical drainage was performed with release of 180 mL of purulent uterine discharge. With administration of FMOX to treat of endometritis and pelvic peritonitis, the amount of vaginal discharge decreased but the fluid collection in the abdominal cavity increased gradually and CRP levels increased to 38.75 mg/dL. Three days after admission, repeat CT revealed a large increase in the amount of the fluid collection in the pelvic cavity and upper abdomen, up to the level of the liver, as well as a collapsed uterine cavity (Fig. 3D and E). Treatment for progressive generalized peritonitis was considered. We selected laparoscopic irrigation and drainage, as minimally invasive interventions, in consideration of her advanced age, poor performance status (PS 4) [1] and possibility of vaginal drainage. Laparoscopic surgery with 2 ports was performed 4 days after admission. Massive purulent ascites and inflammatory adhesions were observed in the entire abdominal cavity (Fig. 4A and B). After extensive suction of the purulent material and irrigation, we found a small scar with serosal spotting on the anterior wall of uterine body, which was attributed to SPP (Fig. 4C). The leakage of purulent uterine discharge into the abdominal cavity might have occurred through this site of perforation. Uterine contractions might have led to scarring by the time of laparoscopic surgery. Hysterectomy or suturing of myometrium was not performed because no bleeding or leakage of purulent discharge from the scar was observed intraopera-

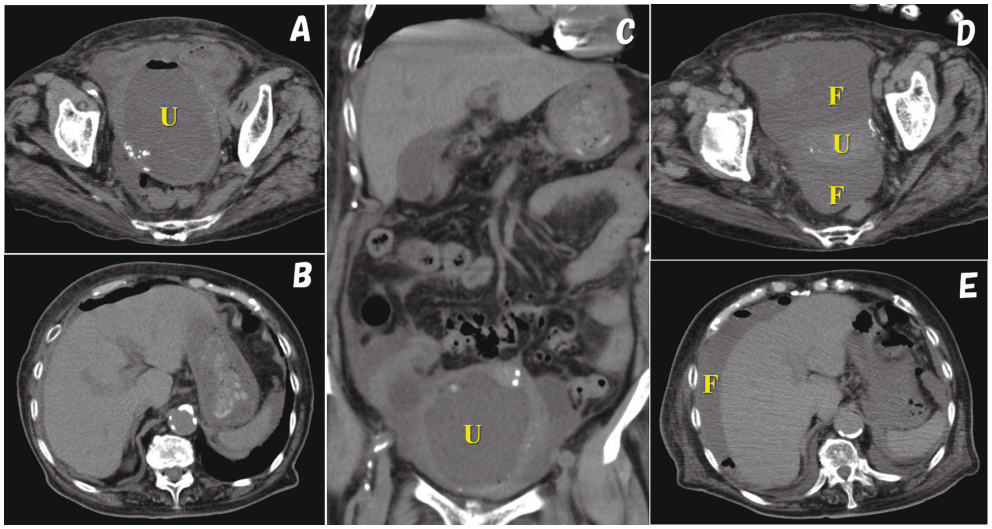

Fig. 3. Preoperative urgent and repeat computed tomography (CT) in Patient 2.
Urgent preoperative CT revealed a fluid collection and free air in the extremely dilated uterine cavity (A and C), although only a small amount of ascites was observed in the pelvic cavity (A, B, and C). Repeat CT revealed a collapsed uterine cavity and a large fluid collection in the pelvic cavity (D). The fluid collection came up to the level of the liver (E).

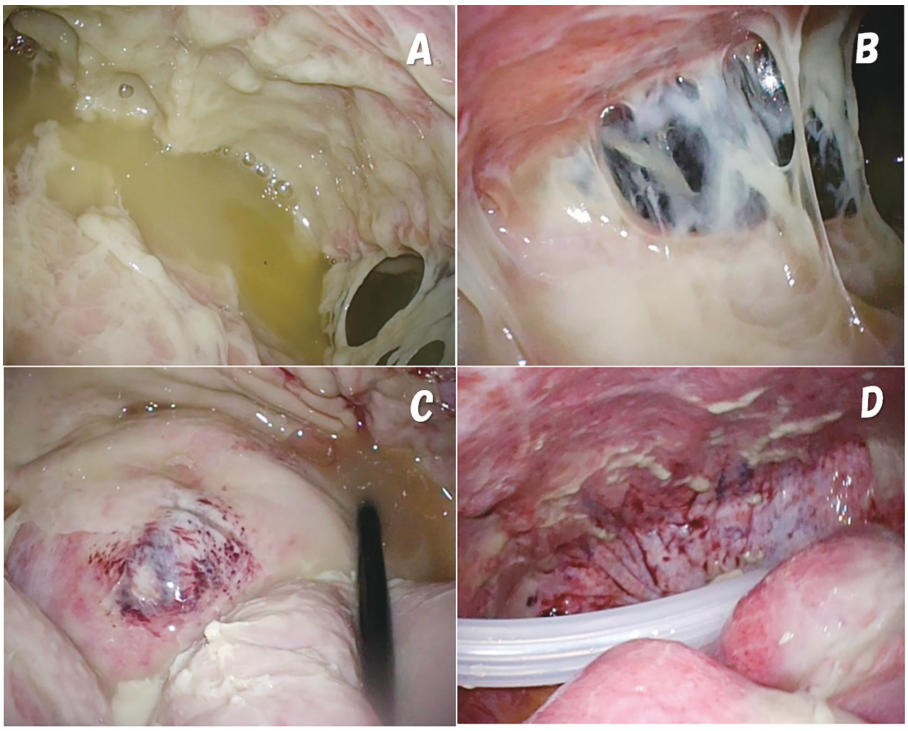

Fig. 4. Findings during laparoscopic surgery in Patient 2.
Massive purulent ascites and inflammatory adhesions were observed in the entire abdominal cavity (A : pelvic cavity, B : upper abdomen). A small scar with serosal spotting was found on the anterior wall of the uterine body (C), which was thought to be the scar of SPP. Leakage of purulent discharge from the uterus into the abdominal cavity might have occurred through this laceration. After suction of the purulent material as far as possible and irrigation of the entire abdominal cavity, a drain was placed in the pouch of Douglas (D).
SPP : spontaneous perforated pyometra.

tively, and transcervical drainage was possible. After placing a drain in the pouch of Douglas (Fig. 4D), bleeding occurred from the surface of liver where adhesiolysis was performed. Hemostasis was accomplished through a small incision in the upper abdomen. The operative time was 2 hours and 40 minutes. Intraoperative blood loss was 914 mL. Most of the operative time and blood loss were related to hemostatic control of bleeding from the surface of the liver. Culture of the peritoneal pus grew *E. coli, ESBL*. Postoperatively, peritonitis decreased with intensive care, including transabdominal and transcervical drainage with administration of MEPM and CLDM, which were selected based on bacterial culture results from the purulent material. The patient's general condition improved ; she was discharged from the hospital, on POD 35.

Discussion

Pyometra, defined as the accumulation of purulent material in the uterine cavity resulting from interference with natural drainage via the uterine cervix, is an uncommon condition that mainly occurs in postmenopausal women. It is reported to occur in 0.1–0.5% of all gynecologic patients and in 13.6% of elderly gynecologic patients (over 60 years of age)[2–7]. This condition, including cervical stenosis, is commonly caused by benign or malignant gynecologic tumors, atrophic cervicitis with aging, puerperal infection, radiation cervicitis, long-term use of an intrauterine device, or congenital uterine anomalies[5,6,8]. In general, the most common microorganisms causing pyometra are *E. coli, Streptococcus spp., Bacteroides fragilis,* and *Peptostreptococcus anaerobius*[7,9]. The main symptoms of pyometra are lower abdominal pain, purulent vaginal discharge, and postmenopausal bleeding, although more than 50% of patients are asymptomatic.

SPP is very rare ; it is estimated to account for less than 5% of all cases of pyometra[2,8–11]. Although abdominal pain, vomiting, and fever predominate as the presenting symptoms, there are no symptoms specifically associated with SPP, often making accurate diagnosis difficult[11]. Diagnosing pyometra is relatively easy based on imaging studies such as ultrasonography or CT. Findings include uterine enlargement and dilation of the uterine cavi-

ty with accumulated purulent discharge and free air. Once perforation has occurred, preoperative diagnosis of pyometra becomes more difficult because of the collapse of the uterine cavity[7]. Since most cases of SPP lead to a fluid collection (purulent discharge) and free air in the abdominal cavity, sometimes it can be misdiagnosed as GI tract perforation. With radiological findings of pneumoperitoneum, GI tract perforation is the most common preoperative diagnosis because it is the cause of pneumoperitoneum in 85–95% of cases[12]. While CT is often indicated for pneumoperitoneum, the presence of free air in the peritoneal cavity without GI tract perforation can be due to gas-forming microorganisms such as *E. coli* and *Bacteroides fragilis*[11]. Accurate diagnosis of SPP may be still difficult, especially when imaging is reviewed in the emergency setting by non-gynecologists. SPP should be suspected when there is free air in both the peritoneum and the uterine cavity in a patient with generalized peritonitis[2,3]. Gynecologic examination with a speculum and transvaginal ultrasonography, which can occasionally reveal a perforation of the uterine wall, may be the most useful tools for diagnosing this rare disease[5,12,13].

Uno *et al.* reported that an accurate preoperative diagnosis was made in only 8 of 37 patients (21.0%) with SPP that was not associated with malignancy[13]. More than 70% of patients were preoperatively diagnosed as having GI tract perforation (17 cases, 50.0%) or generalized peritonitis (7 cases, 18.4%)[13]. According to a Japanese study by Matsue *et al.* that reviewed 80 cases of SPP, accurate preoperative diagnosis was only made in 20 patients (25%) whereas 44 patients (55%) were preoperatively diagnosed as having GI tract perforation[7]. Although malignant uterine tumor was observed in 15% of all patients with SPP, it was present in 80% (4 of 5 patients) of patients under the age of 60. Thus, the author suggested that uterine malignancy should be suspected in relatively younger patients with SPP[7].

Although the basic treatment of pyometra includes transcervical drainage, irrigation of the uterine cavity, and antibiotics therapy, emergency surgical intervention is required to control generalized peritonitis once SPP occurs. Most previously reported cases of SPP were surgically managed with abdominal hysterectomy and irrigation of the abdominal cavity and drainage, followed by intensive care with administration of broad-spectrum antibiotics postoperatively[5,14,15]. Because most patients with SPP are older and have poor PS, similar to our patients, emergency abdominal hysterectomy might be too invasive given the prolonged operative time and excessive intraoperative blood loss[7]. For patients in poor condition, minimally invasive surgical treatments such as suturing of the perforated myometrium, irrigation of the abdominal cavity, and transabdominal and/or transcervical drainage may be selected as the primary emergency surgical interventions[7]. Laparoscopic surgery provides a less invasive choice, as in Patient 2.

Table 1. Review of the literature on the preoperative diagnosis, surgical intervention, and mortality rate in SPP.

	No of cases (n=60)#	%
Age : mean (range)	72.4 (34–92)	–
Preoperative diagnosis of SPP*	17	28.3
Preoperative diagnosis of GI tract perforation**	25	41.7
With uterine cancer	9	15.0
Mortality : total	16	26.7
with hysterectomy	14/52	26.9
without hysterectomy	2/8	25.0

*SPP : spontaneously perforated pyometra. **GI : gastrointestinal
References 6, 11, 13, and 16–24.

In addition to previous reports[6,11,13], we again reviewed the literature and found 60 cases of SPP with data on preoperative diagnosis, surgical treatment (whether hysterectomy was performed), and prognosis (mortality) to year 2018. The results appear in Table 1. Accurate preoperative diagnosis of SPP was achieved in only 28.3% of SPP cases. Although generalized peritonitis or pneumoperitoneum were the other preoperative diagnoses in patients with SPP, GI tract perforation was the most common preoperative diagnosis (41.7%). The mortality rate was as high as 26.7%. Regarding surgical interventions, 52 cases (86.7%) involved hysterectomy and only 13.3% of patients were treated with drainage, peritoneal irrigation, or suturing of the perforated site without hysterectomy. Mortality in patients with and without hysterectomy was 26.9% and 25.0%, respectively. We also reviewed 96 cases of SPP, including information on the factors mentioned above, from the Japanese literature. Preoperative diagnosis of SPP and GI tract perforation was 32.2% and 51.0% respectively, and the mortality rates of patients with and without hysterectomy were 15.5% and 12.0%, respectively (unpublished data). No significant differences emerge in preoperative diagnosis or mortality rates when comparing publications in English and Japanese. Although

hysterectomy was more commonly selected as a surgical intervention, it might be more invasive for patients of advanced age in poor general condition, which might have worsened the prognosis of these patients. According to the reports of Yildizhan *et al.* and Uno *et al.*, the uterine fundus is the most likely site of perforation (85.7% and 78.8%, respectively) [6,13].

We do not know the actual reasons for the SPP in either case. Enlargement and increased pressure in the uterine cavity or thinning and focal necrosis of the myometrium due to prolonged inflammation might have induced rupture (Case 1) and perforation (Case 2) of the uterine wall. The possibility that stimulation of cervical drainage might have induced perforation of pyometra in Case 2 could not be denied, although the definitive cause is unclear.

In Patient 1, hysterectomy and colectomy, which were performed as primary surgical interventions, might have been too invasive, possibly contributing to the poor prognosis because of prolonged operative time and high blood loss. Since transcervical drainage was not possible due to cervical stenosis, likely due to uterine inflammation in this patient, hysterectomy or suprahysterecomy could not be avoided. However, colectomy might have been postponed and performed as a secondary treatment after the generalized peritonitis resolved. In Patient 2, minimally invasive laparoscopic surgery was selected because transcervical drainage was possible and the site of spontaneous perforation in the uterine body was almost repaired, which might have led to relatively better prognosis.

Conclusion

Here we presented two rare cases of SPP and reviewed the literature focusing on preoperative diagnosis, surgical intervention, and prognosis. Preoperative diagnosis of SPP is difficult. It has a poor prognosis, with a mortality rate as high as 15–27%. Minimally invasive surgery should be favored as the primary intervention if repair of the perforated site and transcervical drainage are possible. Since most patients with SPP are elderly women who occasionally have poor PS, emergency abdominal hysterectomy may be associated with a risk of worsened prognosis in such patients, due to increased surgical invasiveness.

Many cases of SPP were preoperatively diagnosed as GI tract perforation during emergency assessment, due to the acute abdomen accompanied by a peritoneal fluid collection and pneumoperitoneum detected on imaging studies. Gynecologists could be incidentally consulted and required for adequate treatments when a diagnosis of SPP is made during emergency surgery. It is very important for gynecologists to be familiar with SPP and be able to select the most suitable treatment, in consideration of the patient's status and disease severity, especially when they encounter patients with SPP in an emergent setting.

Conflict of interest

There are no conflicts of interest of relevant to this article.

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